CASE REPORT

Teenager with pathological bone fracture secondary to parathyroid adenoma at a tertiary care center in Pakistan

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Abstract:

An excessive production of parathyroid hormone (PTH) by one or more parathyroid glands is known as primary hyperparathyroidism. Patient usually presents with lab results indicating increased levels of calcium and parathyroid hormone, to indeterminate constitutional symptoms such as muscle weakness, memory impairment, hematuria, abdominal pain, constipation, or pathological fractures. Definitive investigation is 99mTc-sestamibi MIBI parathyroid Scintigraphy followed by removal of parathyroid gland. Histopathology is then sent for confirmation of diagnosis.

Case Report:

We present a case report of a 16-year-old teenager presented to surgical clinic with 2-months of constipation, multiple joint pain, difficulty walking and generalized body ache that progressively worsened. Xray Pelvis showed pathological fracture of pubic ramus and femur. Histopathology nodules were composed of clear to eosinophilic cells with round nuclei and compressed parathyroid parenchyma at periphery suggesting parathyroid adenoma.

Keywords: Parathyroid adenoma, primary hyperparathyroidism, pathological bone fracture

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Introduction:

An excessively high PTH along with an elevated ionized calcium level is known as hyperparathyroidism. It belongs to the category of endocrine disorders, which are further divided into primary, secondary, and tertiary. It presents with a single adenoma in 80%, as hyperplasia in up to 15% and less than 1% with parathyroid carcinoma.[1] Parathyroid hormone is produced by the four parathyroid glands. The mechanism of action of Parathyroid hormone is to maintain the serum calcium levels to the basal value, by acting on three different organs of the body. In the bone, the hormone inhibits the osteoblast activity and stimulate osteoclast activity, leading to release of calcium. On the kidney, the hormone increases calcium reabsorption, and blocks phosphate reabsorption from the tubules. The hormone has an indirect action on the gastrointestinal system by acting on the vitamin D receptors.

A variety of signs are indicative of PHPT, ranging from an accidental asymptomatic biochemical finding to vague constitutional symptoms like muscle weakness, memory impairment, hematuria, abdominal pain, constipation, or pathological fractures with lab results showing elevated levels of calcium and parathyroid hormone. It affects approximately one percent of the population, occurring mainly in elderly but in adolescents it is rare.[2]

PHPT is less likely to be seen in the pediatric population with less than five percent of the cases of hypercalcemia in children[3,4] and parathyroidectomy is required for definitive treatment and cure.[5]

Case presentation:

A 16-year-old girl arrived at the surgical outpatient department complaining of two months of constipation, multiple joint pain, and trouble

Table 1: Initial Investigation

LABS	PTH	Calcium	25 Hydroxy Vit-D	Phosphorus	Magnsium	Corrected Calcium	Albumin	Alkalibne Phosphatase
On Admission	1223 pg/mL	13.82mg/dl	62.8ng/Ml	2.1mg/dl	0.70mg/dl	14.98mg/dl	2.84g/dl	15391U/L

Table 2:

LABS	PTH	Calcium	25 Hydroxy Vit-D	Phospho-rus	Alkalibne Phosphatase
On discharge	54.43 pg/ml	8.8mg/dl,	62.8ng/Ml	3.24mg/dl,	91 U/L

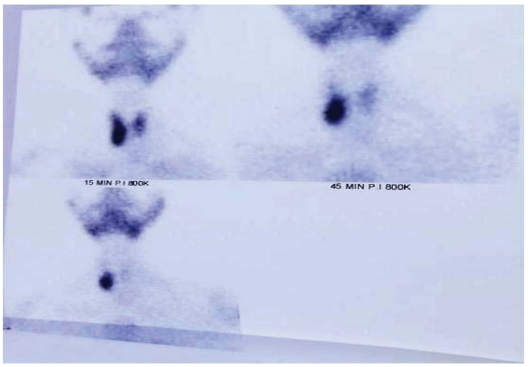

Figure 1 :Sestamibi MIBI Scintigraphy

walking and bearing weight, and generalized body fatigue that progressively worsened. There was no concomitant weight loss, vomiting, fever, or urinary complaint. There was no prior medical hospitalization or fall history. There was no family history of kidney stones, parathyroid illness, or osteoporosis. She initially visited an orthopedic physician for advice because of increasing right hip pain and limited movement. It was discovered upon examination that she had pathological fractures. The orthopedic provided non-operative care such as analgesics, optimum rest, assistive walking, and physical rehabilitation. She was referred to the department of surgery for additional care.In surgery department upon her examination, the right and left hip joints had painful and decreased range of motion on extension and flexion and the patient was unable to walk. In the lower limbs, there was noticeable mild muscular atrophy but normal symmetry. Further systemic examination including abdominal and neck examination was unremarkable. X-ray Pelvis showed marked generalized osteopenia, pathological fracture at neck of right femur and inferior pubic ramus of left side. There was irregularity and widening of both sacroiliac joint. This was followed by certain laboratory investigations. Ultrsound KUB and thyroid profile was normal. The results of the biochemical examination at the time.

An initial diagnosis of PHPT was made with Ultrasound of neck showing well defined hypoechoic area measuring 25.1x 9.8mm poster inferior to the right thyroid lobe with vascularity on color doppler. 99mTc-sestamibi MIBI parathyroid Scintigraphy was done that showed normal thyroid at initial static and delayed images with a focal area of increased tracer uptake noted in initial images of lower part of right lobe of thyroid gland that retained activity on subsequent images as shown in Figure 1

Based on history, clinical finding and imaging, diagnosis of PHPT secondary to parathyroid adenoma or hyperplasia was made. An elective surgery under general anesthesia was planned after baseline investigations, anesthesia workup and negative Covid-19 PCR. A minimal invasive parathyroidectomy was done through central approach. Approximately, 2.5x1cm parathyroid gland identified at right lower lobe of thyroid gland and excised. Right recurrent laryngeal nerve was identified and saved. A drop in the plasma PTH level was seen returning into the normal range of 54.43pg/mL at 10 minutes predicting single-gland disease. Serum calcium dropped to 7.7mg/dl following surgery on day 1 and numbness around the mouth was observed 2 days post-operatively. She was treated with calcium gluconate, after which the symptoms were relieved on third post-operative day. All biochemical parameters were improved, and the bone pain was significantly relieved 2 week post-operatively. Her biochemical levels were returned to normal, respectively following surgery as shown in Table 2.

Finally, bone pain completely eliminated, and patient's quality of life significantly improved at 6 months follow up. Histopathology revealed a gross appearance of nodular tissue with cut surface showing tan brown cystic area. Microscopically nodules were composed of clear to eosinophilic cells with round nuclei and compressed parathyroid parenchyma at periphery suggesting parathyroid adenoma.

Discussion:

To attain the best possible surgical result, preoperative localization of the adenoma is essential, and cervical US in conjunction with sesta-MIBI scintigraphy is thought to be the most effective method for defining the anatomic correlates of an adenoma.[6]

Parathyroid adenoma is a well encapsulated benign neoplasm of parathyroid gland which is composed of chief cells and oxyphillic cells. It mostly presents as a solitary glandwith only 5% presentations being double gland.[7] Many patients present with nonspecific symptoms. It is possible that these patients will not always experience the typical signs of PHPT, such as depression, pathologic fractures, and nephrolithiasis. The combined consequences of hypercalcemia and elevated PTH secretion result in the clinical appearance. Increased PTH levels are directly linked to bone pathology and nephrocalcinosis, while vomiting, constipation, dehydration, and an unwillingness to eat are more closely associated with hypercalcemia. Pathological fractures as a presenting feature of primary hyperparathyroidism in young patient with parathyroid adenoma is extremely rare.[8]

Initial management for all patients with symptomatic hypercalcemia is intravenous fluids for excretion of calcium through kidneys. Bisphosphonates, pyrophosphate analogues can be used to decrease bone turnover and increase bone mineralization by inhibiting osteoclast activity but the surgical removal of all diseased parathyroid tissue is the ultimate treatment for primary hyperparathyroidism. Similar case reports have been seen in a young 18 year old female with a single parathyroid adenoma who presented with multiple typical fractures history and underwent surgical excision.[8] Bennett et al. reported a case of an adolescent female, who sustained right femoral neck fracture secondary to parathyroid adenoma.[7]

In the present case we treated a 16-year-old female patient with PHPT caused by parathyroid adenoma through MIP procedure under general anesthesia in a previously asymptomatic female. The diagnosis of parathyroid adenoma was made by following a phased and planned approach for a pathological fracture. After the excision of parathyroid adenoma, 12-months follow-up showed normal biochemical markers.

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Role and contribution of authors:

Uzma Shamim Seth, main idea, manuscript writing, surgery, finalizing.

Muhammad Farooq Umer, discussion writing.

Abdullah Muttaqi, case presentation writing.

Maha Kamal, data retrieval, manuscript writing.

Muhammad Taha Kamal, manuscript writing, finalizing

Syed Ashir Hasnain, introduction writing, data retrieval.

Azizuddin, reference writing, data retrieval.

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